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ORIGINAL ARTICLE

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Centre-level variation in outcomes and treatment for otitis media with effusion and hearing loss and the association of hearing loss with developmental outcomes at ages 5 and 7 years in children with non-syndromic unilateral cleft lip and palate: The Cleft Care UK study. Part 2

A. Hall[1,2] | A. K. Wills[3,4] | O. Mahmoud[5,7] | D. Sell[6] | A. Waylen[4] | S. Grewal[4] | J. R. Sandy[4] | A. R. Ness[3,4]

[1]Life and Health Sciences, Aston University, Birmingham, UK

[2]Children's Hearing Centre, St Michael's Hospital, Bristol, UK

[3]National Institute for Health Research (NIHR) Biomedical Research Unit in Nutrition, Diet and Lifestyle at the University Hospitals Bristol NHS Foundation Trust and the University of Bristol, Bristol, UK

[4]Bristol Dental School, University of Bristol, Bristol, UK

[5]School of Social and Community Medicine, University of Bristol, Clifton, Bristol, UK

[6]North Thames Regional Cleft Service, Speech and Language Therapy Department and Centre for Outcomes and Experience Research in Children's Health, Illness and Disability (ORCHID), Great Ormond Street Hospital NHS Foundation Trust, London, UK

[7]Department of Applied Statistics, Helwan University, Cairo, Egypt

Correspondence
Jonathan R. Sandy, Bristol Dental School, University of Bristol, Bristol, UK.
Email: jonathan.sandy@bristol.ac.uk

Structured Abstract

Objectives: To explore centre-level variation in otitis media with effusion (OME), hearing loss and treatments in children in Cleft Care UK (CCUK) and to examine the association between OME, hearing loss and developmental outcomes at 5 and 7 years.

Setting and Sample Population: Two hundred and sixty-eight 5-year-old British children with non-syndromic unilateral cleft lip and palate (UCLP) recruited to CCUK.

Materials and Methods: Children had air and bone conduction audiometry at age 5. Information on grommet and hearing aid treatment was obtained from parental questionnaire and medical notes. Hearing loss at age 5 was defined as >20 dB in the better ear and history of OME and hearing loss was determined from past treatment. Children with sensorineural hearing loss were excluded. Associations were examined with speech, behaviour and self-confidence at age 5 and educational attainment at age 7. Centre variation was examined using hierarchical models and associations between hearing variables and developmental outcomes were examined using logistic regression.

Results: There was centre-level variation in early grommet placement (variance partition coefficient (VPC) 18%, *P*=.001) and fitting of hearing aids (VPC 8%, *P*=.03). A history of OME and hearing loss was associated with poor intelligibility of speech (adjusted odds ratio=2.87, 95% CI 1.42-5.77) and aspects of educational attainment.

Conclusions: Hearing loss is an important determinant of poor speech and treatment variation across centres suggest management of OME and hearing loss could be improved.

KEYWORDS

centralization, developmental outcomes, hearing loss, unilateral cleft lip and palate

1 | INTRODUCTION

Otitis media with effusion (OME) is prevalent in young children with cleft palate.[1,2] In children with cleft palate, OME is commonly associated with conductive hearing loss, which is typically more severe than in children without cleft palate.[1,3] OME related conductive hearing loss influences the consistency and stability of the afferent auditory signal, which can potentially compromise phonological development and attention for listening.[4] The National Institute for Health and Care Excellence (NICE) guidelines[5] for managing OME in children with cleft palate in the UK state that management should be undertaken by local Ear, Nose and Throat (ENT)/audiology services in liaison with the regional cleft lip and palate team, and that grommets or hearing aids are possible treatment options for those with OME and persistent hearing loss.

Our previous research examined the impact of centralization on treatment for OME and hearing loss, and hearing outcomes in 5-year-old children with unilateral cleft lip and palate (UCLP) in the Cleft Care UK study.[6] Since centralization, the use of grommets has reduced and the use of hearing aids has increased.[6] However, there was no difference in the amount of hearing loss at age 5 for children following centralization of services with approximately 50% of children having abnormal middle ear status.[6] Previous studies have suggested that grommet use for children varies geographically[7,8] with an 8.5-fold variation across local authorities in England over the period 2011-2012. We are not aware of any studies that have examined centre-level variation in treatment or outcomes for hearing in children with cleft lip and palate.

While hearing ability at a point in time is important, the impact of OME on child development depends on the duration of any associated hearing loss and the timing during sensitive periods of development.[4] For otherwise typically developing children with no additional risk factors, systematic reviews and meta-analyses of prospective and intervention studies have concluded there is little to no long term impact of OME on language[9,10] or other developmental outcomes such as behaviour or quality of life.[11] However, many of the included studies do not account for the presence, severity or timing of hearing loss.

For children with cleft palate, who have a higher risk of persistent OME and hearing loss as well as for developing a speech disorder, the impact of OME on development may be greater than for other children.[12,13] However, there has been little research in this area and children with cleft palate are often excluded from studies. A systematic review examining the effectiveness of grommets for children with cleft palate and OME[14] identified three studies focusing on speech and language outcomes, of which only the study by Hubbard et al.[15] was identified as high quality. Hubbard et al.[15] compared two cohorts of children in the US receiving early versus delayed treatment for OME and found poorer speech articulation in the group receiving delayed treatment. There has been little research on OME, hearing loss and their association with other areas of development.

In this paper, analysis of Cleft Care UK is extended to explore centre-level variation in OME and hearing loss (and associated treatments) within this centralized multidisciplinary service and to examine the association between OME and hearing loss and a range of developmental outcomes at 5 and 7 years.

2 | METHODS

2.1 | Study sample

Data from CCUK were used. This is a UK-wide cross-sectional study of 5-year-old children born between April 2005 and March 2007 with unilateral cleft lip and palate (UCLP). A full description of recruitment procedures and eligibility criteria can be found elsewhere.[16] Briefly, of 359 eligible children, consent for participation was obtained from 268 (75%) children and parents. Ethical approval was obtained (REC reference number: 10/H0107/33, South West 5 REC). Children were excluded if they had confirmed or possible sensorineural hearing loss based on audiometry at age 5 (n=8).

2.2 | OME and hearing treatment and outcome measures

The hearing measures used in CCUK have been described in detail elsewhere.[6] In brief, children had a full audiological assessment at age 5 including air and bone conduction audiometry. Average hearing threshold level was calculated across the frequencies 0.5-4 kHz in each ear. The audiological and ENT treatment history was obtained through parental questionnaire and from medical notes, including information about grommet and hearing aid treatment.

For developmental outcomes, the OME and hearing loss exposure was examined in two ways. Firstly, *history of OME and hearing loss* was determined from a child's treatment history (there were no early audiological records available through CCUK). This was defined as ever having worn hearing aids, or having grommets or t-tubes inserted (apart from when fitted simultaneously with palate closure) up to age 5. Secondly, *hearing loss at age 5* was determined as thresholds >20 dB in the better hearing ear (binary variable). For the treatment indicators, data were classified according to whether a grommet had been inserted, whether grommets were fitted at palate closure and whether hearing aids were fitted.

2.3 | Developmental outcomes

A range of developmental outcomes were examined, selected based on core outcomes identified by parents and clinicians as important for cleft palate and OME research[17]: speech, behaviour and self-confidence measured at the age 5 research clinic and educational attainment at age 7 obtained from linkage to educational records.

2.4 | Speech measures

Children's speech was assessed at age 5 using speech audio-video recordings taken by specialist speech and language therapists. Two independent listeners undertook perceptual analysis using the CAPS-A tool to give a structural score (derived from measures of hypernasality,

audible nasal emission, nasal turbulence and the passive category), and articulation measure (derived from the anterior, posterior and non-oral categories). There were four possible categories for the structural and articulation scores, from 0 (unaffected) to 3 (severely affected). A measure of speech intelligibility/distinctiveness was also derived, which is a measure of the ability of an unfamiliar listener to understand speech. Further details of the derivation of these scores are given in Sell et al.[18] (within this supplement) and Sell et al.[19]

2.5 | Child behaviour and self-confidence

Behaviour of the child was based on parental assessment using the Strengths and Difficulties Questionnaire.[20] Scores examined were the hyperactivity subscore and the total difficulties score, as identified from previous cohort studies of behavioural sequelae of OME.[21] Higher scores represent more hyperactivity behaviour or greater behavioural difficulties and the data for both scores were split into tertiles; more details on these outcome measures are reported in Waylen et al.[22] (within this supplement). Self-confidence of the child was based on parental response to the question "Do you feel your child's self-confidence has been affected by the cleft?" Responses could be scored between 1 to 10 where 1 represents as negative effect of the cleft on self-confidence, 5 is no difference and 10 represents a positive effect. Scores were grouped so scores from 0 to 3 were categorized as a negative effect of the cleft and compared to the remaining reference category.

2.6 | Educational measures

Key stage 1 education standardized attainment tests (SATs), in reading, writing, speaking and listening, maths and science were used as measures of educational attainment at age 7. Tests are scored in the following categories: W, 1, 2 (with fine levels C, B and A) and 3, where W is the lowest and 3 is the highest. Speaking and listening and science are not scored at the fine levels. W and 1 categories were combined for analysis due to small cell counts.

2.7 | Confounders, mediators and effect-moderators

Confounders examined were age and gender of the child and socioeconomic status (SES) of the family, as these factors are associated with prevalence of hearing loss and developmental outcomes.

The Index of Multiple Deprivation was used as a proxy of SES. This is a geographically based (postcode) relative measure of deprivation and consists of a weighted score covering up to seven domains (income, employment, education, skills and training, health and disability, crime, housing and living environment). Higher scores indicate higher deprivation. The score is used to rank neighbourhoods from most deprived to least deprived. Deprivation ranks were obtained from England (http://geoconvert.mimas.ac.uk/help/imd-2007-manual.pdf), Scotland (http://www.gov.scot/Topics/Statistics/SIMD/SIMDPostcodeLookup/ScotlandPostcodeLookup) and Wales (https://statswales.gov.wales/Catalogue/Community-Safety-and-Social-Inclusion/Welsh-Index-of-Multiple-Deprivation/Archive/WIMD-2011). These neighbourhood ranks are subject to small changes over time and IMD scores go back to 2007, 2009, and 2011 for England, Scotland and Wales, respectively. Ranks were used from these years as they are closest to the year of birth and to the birth to 5-year exposure period of our cohort. The ranks are relative to other neighbourhoods within each country, they are therefore not comparable on an absolute scale between countries. To harmonize, individuals were classified in the lowest quartile within our cohort for each country as living in the most deprived areas.

The IMD score was also examined as a potential effect modifier of any associations. SES is associated with many developmental outcomes,[23] and in accordance with the cumulative risk model of OME and development[24,25] it was hypothesized that children from more disadvantaged backgrounds may be more susceptible to the impact of a history of hearing loss than similarly hearing impaired children from more privileged backgrounds. For analyses examining the history of OME and hearing loss, hearing loss at age 5 was examined as a potential mediator to determine whether any observed associations were explained by current hearing levels rather than previous history of hearing loss.

2.8 | Statistical analysis—centre-level variation

Centre-level variation in ENT and audiological treatment (grommets and hearing aids), history of OME and hearing loss, hearing threshold levels and hearing loss at age 5 was examined using hierarchical regression. Based on these models, we estimated the variance partition coefficient (VPC)—a measure of the proportion of total variation that can be attributed to centre, and used estimates from the model to predict the mean outcomes in each centre. Likelihood ratio tests were performed to assess whether any observed variation between centres could be attributed to chance. All results are adjusted for differences in age and sex. Full details of the method for examining centre-level variation is described in Wills et al.[26] (within this supplement).

2.9 | Statistical analysis—OME, hearing loss and developmental outcomes

Odds ratios were estimated to assess the association between the hearing exposures and outcomes. For binary outcomes (self-confidence), logistic regression was used. For the ordinal outcomes, ordered proportional odds logistic regression was used. Four sets of models were fitted for each outcome and exposure. First a minimally adjusted model including age and sex (model a); second, model (a) with additional adjustment for SES (model b); thirdly model (b) with additional adjustment for hearing loss at age 5 as a potential mediator (model c). Finally models including an interaction term between each exposure and the deprivation index were fitted to test whether children from more disadvantaged backgrounds are more susceptible to the impact of a history of hearing loss (model d). Given the relatively small sample size, we also report results stratified by SES if the *P*-value from the interaction test was <0.1. For the hearing loss exposure at

age 5, only models (a), (b) and (d) were fitted. Analyses were run using all available data and restricting the sample to complete data. The findings were unchanged and so the analysis from all available data is presented in this report.

3 | RESULTS

3.1 | Sample description

Table 1 describes the analysis sample. Of 268 children who were recruited into the study, eight had confirmed or possible sensorineural hearing loss and were excluded from the sample. Two hundred and ten (78%) had at least one outcome measure and exposure measure and thus formed part of the analysis. Approximately 2/3 of the sample were boys, and the mean age was 5.6 years. The average deprivation score of the group was lower than the median in England, Scotland and Wales. Forty-five per cent (n=89/197) of the study sample had a history of OME and hearing loss, having received either grommets or hearing aids by age 5, and 20% had hearing loss in the better hearing ear at age 5 years.

3.2 | Centre-level variation in treatment and hearing outcomes

Table 2 reports the results of the between-centre variability analysis for the ENT and audiological treatment variables and outcomes. There was some evidence that the prescription of hearing aids and rates of grommet insertion at palate closure differed by centre. Approximately 18% and 8% of the total variation in these treatments, respectively, could be assigned to differences between centres. The variation in rate of grommet insertion at palate closure was mainly reflected by two centres with rates above 40% (Figure 1), this compares to the rate in the average centre of 8%. The variation in hearing aid fitting was driven by one particular centre that had a rate of 26%, all other centres had fitting rates below 13% (Figure 2) and the predicted proportion in the so-called average centre was 8%. There was no centre variation in grommet insertion rate, hearing levels or hearing loss at age 5 or history of OME and hearing loss (see Figures 3-6).

3.3 | History of OME, hearing loss and developmental outcomes

Table 3 shows the associations of a history of OME and hearing loss with each developmental outcome. There was evidence of associations with all measures of speech at age 5 and with educational attainment tests for speaking and listening. The associations with speech intelligibility and articulation remained after further adjustment for deprivation score but were less convincing for the speech structural score. Of particular note, was the finding that children with a history of OME and hearing loss had almost three times the odds of having poorer speech intelligibility, and over twice the odds of articulation disorder, compared to those with a negative history. These associations were not mediated by hearing loss at age 5. There was no

TABLE 1 Description of analysis sample[a]. Results are n (%) unless stated

Variable	N	n (%)
Gender, Boys	210	141 (67.1%)
Age (years), Mean (SD)	210	5.61 (0.39)
Deprivation score (percentile), Median (IQR)	189	41 (18, 68)
History of OME and hearing loss(yes)	197	89 (45.2%)
Hearing loss in the better hearing ear at age 5 (>20 dB)	204	43 (21.1%)
Speech[b]		
Intelligibility		
0		108 (58.4%)
1	185	14 (7.6%)
2		34 (18.4%)
3 or 4		29 (15.7%)
Structure		
0		115 (60.8%)
1	189	34 (18.0%)
2		1 (0.5%)
3		39 (20.6%)
Articulation		
0		122 (63.2%)
1	193	17 (8.8%)
2		29 (15.0%)
3		25 (13.0%)
Behavioural[c]		
Hyperactivity(/10), Median (IQR)	164	4 (2, 6)
Goodman total score (/40), Median (IQR)	160	9 (5.5, 12)
Psychology[d]		
Self-confidence(/10), Median (IQR)	194	5 (5, 5)
Education Key Stage 1[e]		
Speaking & listening		
1		26 (16.8%)
2	155	102 (65.8%)
3		27 (17.4%)
Reading		
1		22 (14.2%)
2C		23 (15.8%)
2B	155	29 (18.7%)
2A		43 (27.7%)
3		38 (24.5%)
Writing		
1		28 (18.1%)
2C		37 (23.9%)

(Continues)

TABLE 1 (Continued)

Variable	N	n (%)
2B	155	39 (25.2%)
2A		24 (15.5%)
3		27 (17.4%)
Maths		
1		15 (9.7%)
2C		31 (20.0%)
2B	155	39 (25.2%)
2A		34 (21.9%)
3		36 (23.2%)
Science		
1		17 (11.0%)
2	155	108 (69.7%)
3		30 (19.4%)

[a]Based on data from all eligible children (n=260) that had at least one measured outcome and exposure variable (n=210).
[b]Higher scores indicate poorer speech.
[c]Higher scores indicate more behavioural difficulties.
[d]A score of 1 indicates a negative effect of the cleft on self-confidence, 5 indicates no difference and 10 a positive effect.
[e]Higher scores indicate higher educational attainment.

evidence of associations with the behavioural or psychological outcomes or any of the other education key stage outcomes.

3.4 | Hearing loss at age 5 and developmental outcomes

Table 4 shows the associations between hearing loss at age 5 and the developmental outcomes. There was no evidence of any association with the age 5 developmental outcomes we examined. However, there was consistent evidence of an association between hearing loss at age 5 and all of the educational outcomes at 7 years among children living in the most deprived quartile in our cohort. Table 5 shows these interactions as well as reporting the odds ratios stratified by living in the most deprived quartile or not. There was no association between hearing loss at age 5 and any of the educational scores among children outside the bottom quartile of deprivation. However, among children living in the most deprived areas, those with hearing loss were much more likely to have poorer educational test scores. The increase in odds of a poorer outcome if a child had hearing loss ranged from four- to eleven-fold, although the confidence intervals were wide reflecting the small sample size and uncertainty in these estimates.

Table 5 also shows that there was an interaction between SES and the history of OME and hearing loss exposure for the mathematics educational score. Again, the association of hearing loss with educational outcome was only present among the children living in the most deprived area. However, the evidence for this interaction was weak (P=.09), it was also the only statistically suggestive interaction out of 11 examined for this exposure and was inconsistent with the results of the interaction tests for the other educational outcomes.

To add context to these interactions, there was no evidence of an association between deprivation and hearing loss (P=.7) or deprivation and history of OME and hearing loss.

4 | DISCUSSION

There was variation in the fitting of hearing aids and grommet insertion at palate closure across centres within this centralized multidisciplinary service but no variation in hearing status at age 5. Children with a history of OME and hearing loss had poorer speech at age 5 than children with no history of hearing loss. There was weaker evidence that OME and hearing loss history was associated with the listening and speaking educational attainment test at age 7. Furthermore, there is a suggestion of an association between hearing loss at age 5 and educational attainment at age 7 in children living in more disadvantaged areas but not in other areas.

TABLE 2 Predicted mean with each hearing treatment and outcome for the so-called average centre and the between-centre variability

Outcome	n	Predicted proportion unless stated (95% CI)	VPC	P-value[a]
Grommets inserted (Yes)	195	0.39 (0.24, 0.57)	0.03	.9
Grommets at palate closure (Yes)	149	0.08 (0.01, 0.55	0.18	<.001
Fitted hearing aid (Yes)	198	0.08 (0.02, 0.32)	0.08	.03
Best hearing threshold levels (mean dB)	193	14.2 (11.6, 16.9)	0.04	.26
Worst hearing threshold levels (mean dB)	193	22.6 (19.2, 25.9)	0.02	.9
Current best hearing >20 dB (Yes)	204[†]	0.20 (0.11, 0.32)	0.02	.5
History of OME/hearing loss (Yes)	197[†]	0.41 (0.25, 0.60)	0.03	.9

VPC, variance partition coefficient.
All results are adjusted for age and sex.
[a]A test of the null hypothesis that there is no between-centre variation.

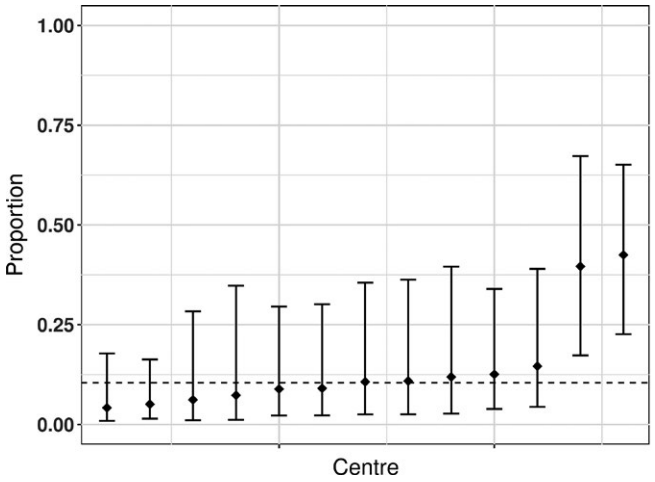

FIGURE 1 Predicted proportion of children with grommets inserted at palate closure in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

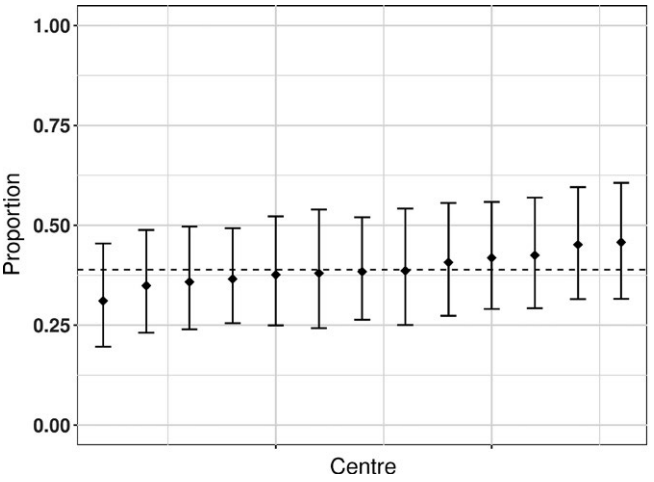

FIGURE 3 Predicted proportion of children with grommets inserted in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

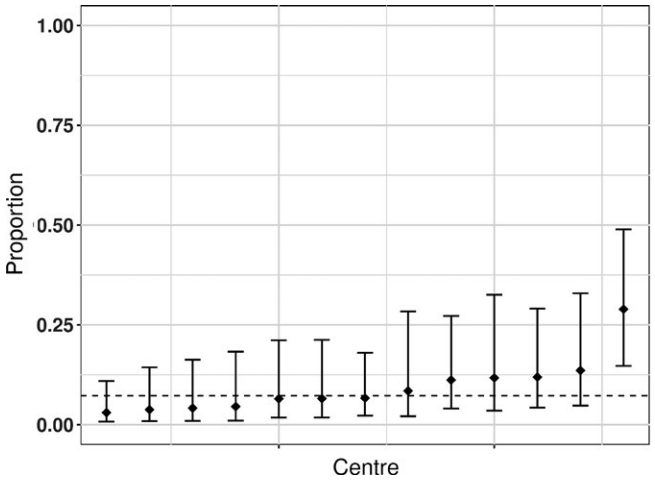

FIGURE 2 Predicted proportion of children with a fitted hearing aid in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

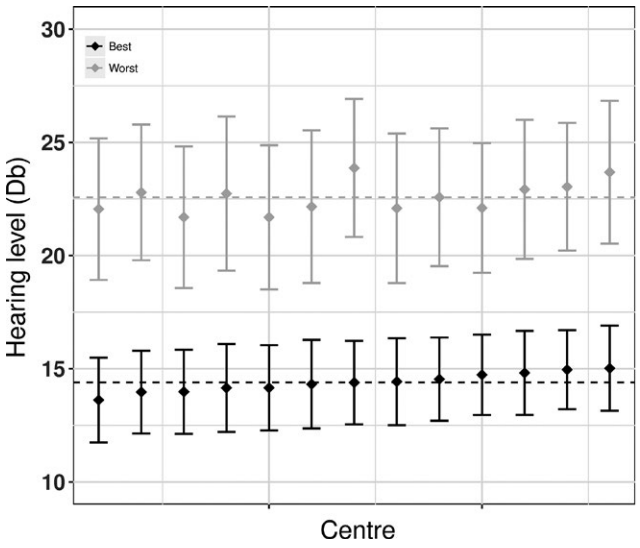

FIGURE 4 Predicted mean hearing levels (dB) in best and worst ear in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

4.1 | Centre-level variation in treatment

The presence of variation in hearing aid fitting is likely to reflect clinician preference in treatment decision-making for managing OME and hearing loss. The fitting of more hearing aids in some centres may indicate clinicians with a preference for hearing aids over grommets. Such an approach was advocated by Maheshwar et al.[27] who described their centre's approach to managing OME and hearing loss in children with cleft palate as "non-interventionist" relying predominantly on the provision of hearing aids with grommets used only in limited circumstances. Although the National Institute for Health and Care Excellence[5] recommends both grommets and hearing aids as suitable interventions for OME and hearing loss, both of which have associated risks and benefits, there is uncertainty among clinicians about the best strategy for managing OME in children with cleft palate.[28] The centres with higher rates of hearing aid fitting could be those that share treatment decision-making with parents, and it is the parents rather than clinicians who are choosing hearing aids over grommets. The NHS have supported development of a patient decision aid to assist patients with decision-making about OME[29] and there is evidence for a range of health conditions that when patients are informed and supported with shared decision-making they are less likely to choose surgery as a treatment option.[30] However Tierney et al.,[31] in a study of parental experiences of managing OME in their child with cleft palate, found that most parents did not recall being offered hearing

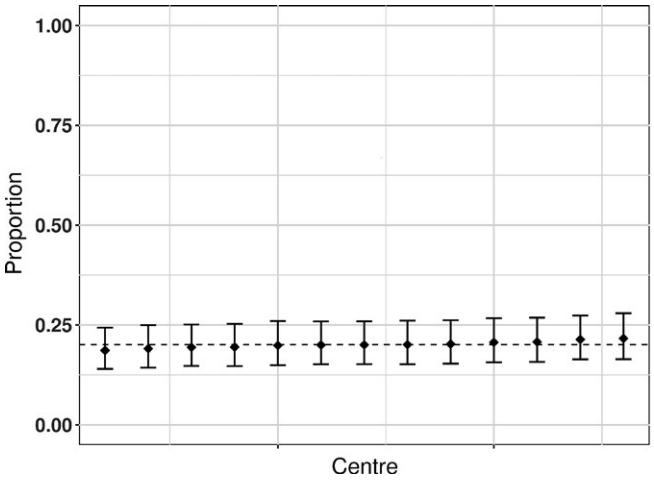

FIGURE 5 Predicted proportion of children with best hearing levels >20 dB art age 5 in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

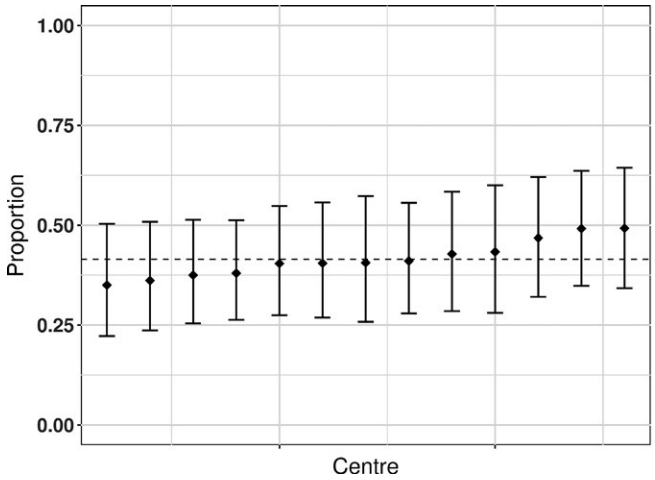

FIGURE 6 Predicted proportion of children with a history of OME or hearing loss in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

aids as an initial treatment option, suggesting that shared decision-making for OME is not widespread. The regular or routine placement of grommets at the time of palate closure was advocated in the past but is now more contentious. The National Institute for Health and Care Excellence[5] state that insertion of grommets at palate closure is not recommended as routine practice and should only be completed after careful otological and audiological assessment. A recent clinician survey indicated that it is not current practice to insert grommets at palate repair[28] and this is consistent with our findings with the practice only occurring at higher rates in two centres.

4.2 | OME and hearing loss and speech

The association between history of OME and hearing loss and speech are consistent with the small number of studies in the literature examining the association between hearing and speech in

TABLE 3 Associations[a] (odds ratios—OR) between history of otitis media with effusion (OME) and hearing loss (hearing aids or grommets) and developmental outcomes

	Model (a)[b]			Model (b)[b]			Model (c)[b]		
Outcome	**N**	**OR (95% CI)**	***P***	**N**	**OR (95% CI)**	***P***	**N**	**OR (95% CI)**	***P***
Speech									
Intelligibility	173	1.96 (1.10, 3.50)	.023	144	2.47 (1.28, 4.78)	.007	135	2.87 (1.42, 5.77)	.003
VPD	176	1.82 (1.01, 3.28)	.047	146	1.58 (0.82, 3.05)	.18	137	1.53 (0.75, 3.12)	.24
Articulation	180	1.78 (0.99, 3.21)	.056	150	2.11 (1.09, 4.09)	.027	140	2.37 (1.17, 4.82)	.017
Behavioural									
Hyperactivity	153	1.48 (0.82, 2.68)	.19	128	1.28 (0.67, 2.48)	.46	120	1.28 (0.64, 2.55)	.48
Total score	149	1.17 (0.65, 2.12)	.6	125	1.15 (0.59, 2.22)	.7	122	1.03 (0.53, 2.02)	.9
Psychology									
Self-confidence	184	0.86 (0.29, 2.53)	.8	153	1.16 (0.37, 3.61)	.8	144	1.28 (0.40, 4.07)	.7
Education key stage 1									
Speaking & listening	147	2.02 (1.01, 4.07)	.048	142	1.94 (0.94, 4.00)	.071	132	1.83 (0.84, 4.00)	.13
Reading	147	1.21 (0.68, 2.17)	.5	142	1.12 (0.62, 2.04)	.7	132	1.08 (0.57, 2.04)	.82
Writing	147	1.30 (0.73, 2.34)	.37	142	1.18 (0.65, 2.16)	.6	132	1.14 (0.60, 2.16)	.7
Maths	147	1.43 (0.80, 2.55)	.23	142	1.39 (0.76, 2.52)	.28	132	1.41 (0.75, 2.66)	.29
Science	147	1.48 (0.73, 3.03)	.28	142	1.37 (0.65, 2.88)	.41	132	1.32 (0.59, 2.92)	.5

[a]Outcomes were coded such that the OR captures the odds of a poorer outcome in the exposed (history of OME and hearing loss) v unexposed categories.

[b]Model (a): adjusted for age and sex; model (b): adjusted for age, sex and SES; model (c): adjusted for age, sex, SES and hearing loss at age 5.

TABLE 4 Associations[a] (odds ratios—OR) between hearing loss at age 5 (better ear>20 dB) and developmental outcomes

	Model (a)[b]			Model (b)[b]		
Outcome	**N**	**OR (95% CI)**	***P***	**N**	**OR (95% CI)**	***P***
Speech						
Intelligibility	180	1.07 (0.50, 2.28)	.867	152	1.16 (0.50, 2.72)	.729
VPD	183	1.00 (0.47, 2.11)	.999	155	1.03 (0.44, 2.41)	.944
Articulation	187	1.14 (0.56, 2.32)	.721	159	1.24 (0.56, 2.72)	.592
Behavioural						
Hyperactivity	159	1.46 (0.72, 2.95)	.294	136	1.25 (0.57, 2.73)	.573
Total score	155	1.41 (0.68, 2.90)	.357	133	1.16 (0.52, 2.59)	.72
Psychology						
Self-confidence	189	0.90 (0.24, 3.37)	.874	160	0.97 (0.25, 3.73)	.96
Education key stage 1						
Speaking & listening	149	1.62 (0.68, 3.85)	.276	146	1.61 (0.68, 3.84)	.281
Reading	149	1.30 (0.62, 2.73)	.488	146	1.33 (0.64, 2.77)	.449
Writing	149	1.36 (0.64, 2.88)	0.42	146	1.37 (0.65, 2.86)	0.406
Maths	149	1.09 (0.52, 2.29)	0.821	146	1.04 (0.50, 2.18)	0.922
Science	149	1.81 (0.72, 4.59)	0.21	146	1.80 (0.71, 4.57)	0.216

[a]Outcomes were coded such that the OR captures the odds of a poorer outcome in the exposed (hearing loss at age 5 years) v unexposed categories.
[b]Model (a): adjusted for age and sex; model (b): adjusted for age, sex and SES.

children with cleft palate. Observational studies comparing speech development in children with cleft palate, with and without a history of hearing loss indicated that OME and hearing loss was associated with early speech production difficulties,[32] articulation errors[33,34] and poorer speech intelligibility.[34] Observational studies comparing early with late treatment for OME in children with cleft palate found an advantage of early treatment on speech articulation and need for speech therapy, but not nasal resonance.[15] The association with speech articulation rather than structure suggests the association is causal although a common structural anomaly causing both the speech disorder and increasing likelihood of OME[35] cannot be ruled out. If the association is causal there are three ways that OME could disrupt speech development in children.[36] Firstly, hearing loss from OME typically fluctuates,[37] which could lead to an inconsistent auditory representation of speech sounds. Secondly a persistent reduction in audibility could lead to reduced speech discrimination ability, where increasing hearing loss and hence reduced perception of phonemic cues is associated with poorer speech production.[38] A mild to moderate hearing loss from OME will have the greatest effect on the audibility of high frequency consonants in speech, the speech sounds most likely to be affected in children with cleft palate. Finally reduced audibility of sound could lead to lower attention for listening. For typically developing children, the findings of studies examining speech production in children with OME are mixed; detailed small scale qualitative studies report articulation problems in the speech of children with OME and hearing loss, whereas larger scale studies using less detailed speech measures typically find no association of OME with speech problems, although not always accounting for hearing loss.[24] However a recent large scale longitudinal population study examining risk factors for persistent speech sound disorder which was classified based on detailed analysis of speech samples showed that children with a history of grommets had twice the odds of having speech disorder at age 8.[39] The observation that history of OME and hearing loss was most strongly associated with intelligibility indicates the potential importance of hearing loss on the development of speech. Although intelligibility as a measure is controversial due to the influence of characteristics of the speaker unrelated to the cleft and limitations of the rating scale (reviewed in Sell and Pereira)[40] the findings indicate hearing loss is having a detrimental effect on a child's ability to be understood. This resonates with the findings of the qualitative study by Tierney et al.[41] on the impact of OME in children with cleft palate: social interactions were identified as a key theme, with OME and hearing loss described as negatively affecting a child's social interactions with peers and their participation in activities.

4.3 | OME and hearing loss and other developmental outcomes

There was some suggestion that an early history of OME and hearing loss was associated with poorer scores on the speaking and listening educational attainment tests at age 7, suggesting the impact of hearing on speech measured at age 5 continues to have a detrimental effect beyond this age. A number of studies have reported poorer educational attainment in children with cleft palate compared to unaffected controls,[42-44] but there have been few studies examining the

TABLE 5 Deprivation stratified associations (odds ratios—OR)[a] of a history of otitis media with effusion and hearing loss, and hearing loss at age 5 yr with developmental outcomes

	Least deprived (>25th percentile)		Most deprived (<25th percentile)		
	OR (95% CI)	*P*	OR (95% CI)	*P*	*P* (interaction)[b]
History of OME & hearing loss					
Maths	1.08 (0.55, 2.12)	.8	3.71 (1.04, 13.22)	.043	.09
Hearing loss at age 5 (better ear>20 dB)					
Speaking & listening	0.87 (0.32, 2.38)	.8	11.3 (1.84, 69.4)	.009	.016
Reading	0.82 (0.36, 1.9)	.4	10.4 (1.7, 63.1)	.011	.012
Writing	0.82 (0.36, 1.93)	.7	10.1 (1.7, 59.4)	.01	.013
Maths	0.68 (0.29, 1.60)	.38	4.8 (1.03, 22.8)	.046	.031
Science	1.13 (0.39, 3.31)	.8	7.18 (1.28, 40.2)	.025	.07

[a]Outcomes were coded such that the OR captures the odds of a poorer outcome in the exposed v unexposed categories. All associations are adjusted for age and sex.
[b]*P*-value for the interaction term—a test of the null hypothesis that the associations are the same in each category of deprivation.

additional impact of hearing loss. Ma et al.[45] found no association of hearing loss with parental reported academic achievement in children with cleft lip/palate in children aged 6-15 years. This should be contrasted with the qualitative findings from parents and children with cleft palate, where a key theme was the detrimental impact of OME on a child's ability to learn and focus in school.[41]

Hearing loss at age 5 was associated with poorer scores on all age 7 education attainment tests, only for children from the most deprived backgrounds. These findings could be due to chance as similar findings were not observed for those exposed to an early history of OME and hearing loss or with the earlier outcome measures. Alternatively they could reflect that hearing loss at school age has a differential effect on development to hearing loss in preschool years and that factors associated with socioeconomic status can protect against its impact. Similar findings have been found in population studies of OME and hearing loss where low socioeconomic status or lack of cognitive stimulation in the home has been associated with a greater impact of OME on cognitive development.[25,46]

It is encouraging that early hearing problems do not appear to have an additional negative impact on a child's psychosocial development or behaviour. There have been a limited number of studies examining the association between hearing loss and other developmental outcomes in children with cleft. Consistent with the findings of this study, Hubbard et al.[15] found no difference in social maturity, self-esteem and behaviour in children with OME and hearing loss treated early versus those treated later. However in the qualitative study of the experience of OME in children with cleft palate by Tierney et al.,[41] emotions were a key theme, with parents and children reporting both negative and positive emotions in relation to OME. The negative emotions included frustration and upset, and were related specifically to the fluctuating and recurrent properties of OME, rather than the severity of the hearing loss; experiences relating to treatment for OME could be positive although children and parents could be anxious about hearing tests and procedures.

4.4 | Strengths and limitations

This study was large (for a study of children with cleft lip and palate), nationwide with a good response rate and a series of validated measures of key outcomes measured with enough precision to demonstrate improvements over time. However, this study does have a number of limitations. First, there was limited power to detect modest centre-level variation in treatment and outcome. Second, children with a history of OME and hearing loss were identified through their treatment history. Audiometric information over the first 5 years was not available to assess the age of onset or severity of OME or to identify children with OME and hearing loss who did not receive treatment. The use of previous treatment with grommets or hearing aids as a proxy measure of persistent hearing loss may also have resulted in the inclusion of children receiving grommets for acute otitis media rather than OME. Children with a history of OME and hearing loss were identified because they received treatment for the condition. It is concerning that poorer speech was observed in the treated group, which, if assuming a causal relationship, implies that grommet or hearing aid treatment alone is not sufficient to overcome development of a speech disorder. This could be because the conductive hearing loss from OME can lead to auditory processing problems, which persist after the hearing loss is resolved[47] and could impact on speech learning. Alternatively, there are other reasons for speech disorders in children with cleft palate, for example structural anomalies of velopharyngeal insufficiency and/or fistulae. Finally, there was no measure of functional listening or language ability to explore how OME and hearing history may relate to language development.

4.5 | Research implications

Future studies should include regular measures of OME and hearing level during early life to examine whether OME and hearing loss

during the early sensitive periods of speech development is worse. These studies need to be larger and longitudinal to investigate the role of hearing across development and should include a measure of language. They should also further examine whether particular groups of children are more at risk of sequelae from OME and hearing loss. Further work is required to develop the optimal early intervention package for managing OME and hearing loss in children with cleft palate, which as well as provision of grommets or hearing aids includes support and interventions for developing speech, listening and communication with accompanying hearing loss, indeed common practice of specialist speech and language therapists in the UK Cleft Centres.

5 | CONCLUSIONS

Children with cleft lip and palate in the UK are now treated by a centralized multidisciplinary service but this has not resulted in improved hearing at age 5. Variations in treatment for hearing loss were evident between centres within this centralized service but there were no variations in hearing level between centres at age 5. OME and hearing loss has an important impact on the development of speech and in particular on intelligibility of speech. Further larger longitudinal studies are required to confirm and extend these findings.

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